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## Accepted Manuscript

Title: Abnormality of standing posture improves in patients with bilateral spastic cerebral palsy following lower limb surgery

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**Abnormality of standing posture improves in patients with bilateral spastic cerebral palsy following lower limb surgery**

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## Highlights

- New index of abnormality of standing posture, Standing profile score, is proposed
- SPS is higher in children with bilateral cerebral palsy than in control subjects
- SPS is reduced after surgery to lower limb
- Reduction in SPS following surgery correlates with reduction in Gait Profile Score

## ABSTRACT

**Objectives:** The degree of abnormality of the gait pattern of children with bilateral spastic cerebral palsy (BSCP) can be reduced by lower limb orthopedic surgery. However, little attention is paid to the effects of surgery on standing posture. Here, we investigated the abnormality of standing posture in young people with BSCP as well as the effects of surgery on standing posture.

**Methods:** We have developed an index of standing posture, the Standing Posture Score (SPS), which is similar in composition to the gait profile score (GPS). We applied SPS retrospectively to 32 typically developing children and 85 children with BSCP before and after surgery to the lower limbs aimed at improving gait. We investigated the relationship between SPS and GPS before surgery and also the relationship between changes in these variables before and after surgery.

**Results:** SPS is significantly higher in young people with BSCP. SPS reduces after surgery and this reduction is correlated with the reduction in GPS.

**Interpretation:** Successful surgery improves the alignment of the lower limbs in BSCP in standing and may have a positive impact on the activities of daily living which depend on a stable and efficient standing posture.

**Keywords:** Bilateral spastic cerebral palsy; Gait profile score; Standing profile score; lower limb orthopaedic surgery

## 1. INTRODUCTION

Cerebral palsy (CP) is the most common cause of physical disability in childhood. It originates from a non-progressive neurological lesion acquired in-utero, perinatally, or during early post-natal development[1,2]. Many children with CP have altered development of the musculo-skeletal system which may lead to abnormal gait patterns and energy inefficiency as a consequence of their neurological deficit [3–5] .

These children may undergo orthopaedic intervention with the aim of improving or slowing the decline of their walking ability. Interventions may include lengthening of musculo-tendinous units, tendon transfers, rotational and displacement osteotomies and stabilization of the hip and foot[6]. A recent systematic review[7] identified numerous retrospective[8–13] and prospective studies[14–19] demonstrating improvements in gait, gross motor function and quality of life following orthopaedic intervention.

3D gait analysis is commonly used to inform surgical decision making[20–22]. A number of indices have been proposed in the literature to measure gait abnormality, including the Gillette Gait Index (GGI)[23], Gait Deviation Index (GDI)[24] and Gait Profile Score (GPS)[25]. The Gait Profile Score (GPS)[25–27] is a well-validated index commonly adopted as an outcome measure for orthopaedic intervention[11,14–17,28]. Thomason et al.[14,15] reported improvements in gait and gross motor function at 1, 2 and 5 years post-operatively using a range of gait indices. Rutz et al.[17] found that patients with greater preoperative gait abnormality had the most significant improvements in GPS, with a mean improvement of 4.3°. In a retrospective study of 40 children with BSCP and

an equinus gait, Firth et al.[11] showed GPS improved by an average of 6.6 degrees 7.5 years after multi-level surgery.

Posture and movement are related, with posture considered a temporary arrested movement[29]. Surgical intervention primarily aims to change the structure of the body with the therapeutic goal of improving the mobility of patients. However, surgical intervention to the lower limbs may also improve standing posture allowing the individual to participate more fully in activities of daily living. No study to date has investigated the effect of surgery on standing posture, and there is no index of standing posture described in the literature.

Our first aim was to develop an index of standing posture, similar to the GPS. We then wished to apply the index retrospectively to data from movement analyses of children with bilateral spastic cerebral palsy (BSCP) conducted before and after orthopaedic intervention to their lower limbs. We hypothesized that children with BSCP would stand in an abnormal posture and that the abnormality of their standing posture, as measured by the index of standing posture, would be related to the abnormality of their gait pattern as measured by GPS. We expected that orthopaedic surgery would reduce the abnormality of standing and that this reduction would be related to the reduction in their GPS.

## **2. METHODS**

In this retrospective study we used data from typically developing (TD) subjects as well as subjects with BSCP. Our analysis was conducted on data from clinical movement

analyses conducted at our laboratory and from TD subjects in our control database.

Each patient had undergone 3D-gait analysis as part of their routine clinical care on two occasions (pre- and post-operatively) using a 9-camera motion analysis system (either a Vicon 612 or ViconGiganet). A standing trial was recorded for approximately 4 seconds. The patient was asked to stand still but not given instructions to adopt any particular posture. At least three representative walking trials were collected at the patient's self-selected walking speed. The markers in each trial were labeled and Vicon software (Plugin Gait, Nexus 1.8.1 software, Vicon Motion Systems Ltd., Oxford, UK) was used to generate joint positions and angles for a complete stride on each lower limb. An identical procedure was followed for the TD subjects.

We developed an index of standing abnormality: the Standing Posture Score (SPS). The index was based on the Gait Profile Score[25] (GPS) but calculated over a period of standing. GPS is calculated from 9 kinematic parameters: pelvic tilt, hip flexion, knee flexion, ankle flexion, pelvic obliquity, hip abduction, pelvic rotation, hip rotation and foot progression. Since there is no direction of travel during standing there was no appropriate reference for the calculation of pelvic rotation and foot progression and these two parameters were excluded. In order to make meaningful statistical comparisons with the GPS, a modified Gait Profile Score (mGPS) was created, which similarly excluded the pelvic rotation and foot progression terms.

All 32 typically developing children aged between 6 and 14 years in our control database were included in this study (8 male, 24 female, mean age 10.1 years, age range 6.4-14.3 years). They were used to calculate a set of reference angles for both standing and walking. For each standing trial an average value across the 32 subjects and across the left and right lower limbs was found for each kinematic variable (the standing

reference angles). For each walking trial, each kinematic parameter was normalised to 51 data points between the initial contact and subsequent initial contact for each lower limb. An average trace (gait reference profile) was created across 3 trials for each subject, across subjects and across the left and right lower limbs.

Data from subjects with BSCP and from TD subjects was analysed in the same way. For each kinematic parameter a standing variable score (SVS) was calculated as the RMS difference between the joint angles for each frame of the standing trial of each subject and the standing reference angles. For each of three walking trials for each subject, gait variable scores (GVS) were calculated as the RMS difference between each of the 51 points of a normalized trial of a subject and the corresponding points of the gait reference profile for each kinematic parameter. Using the same formula as described by Baker et al.[25] for calculating the GPS from the GVSs, the SPS was calculated from the SVSs and the mGPS from the GVSs. The average of the three mGPS scores was calculated.

Data from children from our clinical database were selected if the following criteria were met:

- a) Confirmed diagnosis of BSCP.
- b) GMFCS level I or II.
- c) Aged 4 - 20 years.
- d) Had pre-operative kinematic analysis up to 2 years before surgery and post-operative kinematic analysis between 0.5 and 3.5 years after surgery.
- e) Successful recording of at least 3 independent barefoot walking trials and an independent standing trial.
- f) No surgery, botulinum toxin injections or serial casting for at least a year prior to the first gait analysis.



The eighty-five young people (55 male, 30 female) with BSCP underwent different combinations of bony and soft tissue operations according to clinical indications as a single event. Timings are given in Table 1. A total of 261 surgeries were performed (average 3.1 procedures per patient) and details are given in supplementary material. The most common procedures performed were hamstring and plantarflexor lengthening and accounted for approximately 80% of the total number of surgical interventions.

Post-operatively each participant engaged in a physiotherapy programme, initially as an inpatient and subsequently in the community.

An unpaired Student's t-test (2-tailed) was used to test for a significant difference in SPS between the TD children and the children with BSCP pre-operatively, and a paired Student's t-test was conducted in the young people with BSCP pre-and post-operatively. Correlations between GPS and SPS were tested for significance using Pearson's product-moment correlation coefficient. The level of significance for all statistical tests was set at  $p < 0.05$ .

### **3. RESULTS**

Participants with BSCP had a significantly greater SPS ( $18.3^\circ \pm 6.8^\circ$ ) than their TD peers ( $6.0^\circ \pm 2.5^\circ$ ) ( $p < 0.001$ ).

Amongst the individuals with BSCP, pre-operative SPS and mGPS were significantly correlated ( $r^2 = 0.35$ ,  $p < 0.001$ ) (Figure 1). Surgery reduced both SPS and mGPS significantly (Table 2) and there was a significant correlation between change in the SPS and change in the mGPS ( $r^2 = 0.38$ ,  $p < 0.001$ ) (Figure 2).

### **4. DISCUSSION**

Our hypotheses were supported by our results. Young people with BSCP had abnormal standing posture (as measured using SPS). We found that in patients with BSCP, who went on to have lower limb surgery, SPS explained 35% of the variation in mGPS. Both abnormality of posture when standing and abnormality of posture when walking significantly improved following surgery. Furthermore, 38% of the change in mGPS could be accounted for by the change in SPS following surgery.

We found an average improvement in mGPS following surgery of 2.5 degrees. Although the mGPS is not directly comparable to the GPS, we found that in our subjects mGPS and GPS were very similar (an average difference of 0.1 degrees). Our improvement in mGPS was smaller than values reported in the literature. Lehtonen et al.[31] found a change of 3.5 degrees five years post operatively and Frith[11] found changes of 5.9 and 6.6 degrees at 12-18 months and 7.5 years after surgery respectively. However both these studies included subjects who were not able to walk independently (GMFCS levels III). Rutz et al. [17] has previously shown that individuals with higher pre-operative GPS values show greater improvements in GPS post-operatively.

Standing in a sustainable and upright position is an important body function. This is illustrated by the inclusion of dimensions requiring standing ability in many of the outcome tools used for the assessment of children with CP. Aspects of the POSNA pediatric musculoskeletal functional health questionnaire [32] reflect the child's ability to stand to perform activities of daily living while the LIFE-H[33] includes questions related to participation, some of which may also require standing ability and the GMFM[34] has a standing dimension. The young people with BSCP in this study had abnormal standing postures, and this may have influenced their mobility and their ability to remain balanced in standing. This may impact their ability to carry out activities of

daily living and their level of participation in peer activities.

This study has shown that abnormality of standing posture and abnormality of walking posture are related. However in our group of subjects who were to go on to have surgical intervention to their lower limbs, abnormality of standing posture only explained about 33% of the variation in abnormality of walking posture. This suggests that there are other significant factors that have not been recorded in this study which influence the relationship. It is possible that large differences in walking and standing posture within the individual may be caused by a disorder of movement, whereas, similarity in walking and standing posture may be caused by a combination of weakness and deformity. Broadly speaking, subjects who have a similar level of standing and walking abnormality (similar SPS and mGPS) may be more appropriate for interventions aimed at realigning the musculoskeleton while subjects who have abnormal walking posture (high mGPS) but standing posture close to normal (lower SPS) may respond better to interventions aimed more at the underlying movement disorder.

This was a retrospective study where we utilised data which had been collected as part of clinical gait analysis. Neither the length of the standing trial or standing orientation was standardized between participants. Over the last two years, we have routinely collected longer trials to evaluate standing balance (40 seconds). To assess whether the short standing trials were representative of longer standing trials we compared SPS from different length extracts from the long trials from 16 subjects with BSCP. We found that there was no significant difference between the mean SPS values calculated on longer or on shorter trials (Single Factor Anova). The average standard deviation of SPS for the different length trials was 0.09 degrees which is approximately 0.5% of the mean value of SPS calculated in our study. This suggests that the short length of standing trial used in

this study did not significantly influence our results.

We also carried out a study to assess the repeatability of our standing trials. We collected two standing trials from 10 subjects (who were not part of our main study, but who fulfilled the criteria for our study but who we saw on a single occasion); one at the start of the data collection session and one at the end. We found an interclass correlation (ICC) of 0.98 and a standard error of measurement (SEM) of 1.05. These results are similar to those of Rasmussen et al.[35] who assessed reliability of the GPS by assessing 18 patients with spastic CP on two occasions within a 10 day period and found an ICC of 0.88 and a SEM of 1.09.

In this study, markers were not reapplied to the subject between the standing trial and walking trials. Any error in marker placement would be present for both the standing and walking trials having the potential to artificially increase the significance of the correlation between the SPS and mGPS. In any prospective study of the relationship between walking and standing posture we would recommend re-application of the markers between standing and walking trials to minimise systematic error.

## **5. CONCLUSION**

The standing posture score (SPS) is a novel index and a useful tool for assessing the abnormality of standing in subjects with cerebral palsy. The abnormality of standing and walking posture both improved after surgery, and a relationship was noted in the degree of change between these two parameters. Analysis of SPS and mGPS may provide insight into the contributions of structural and neurological components of an individual's walking pattern.

## Conflict of interest statement

None of the authors have any conflicts of interest to

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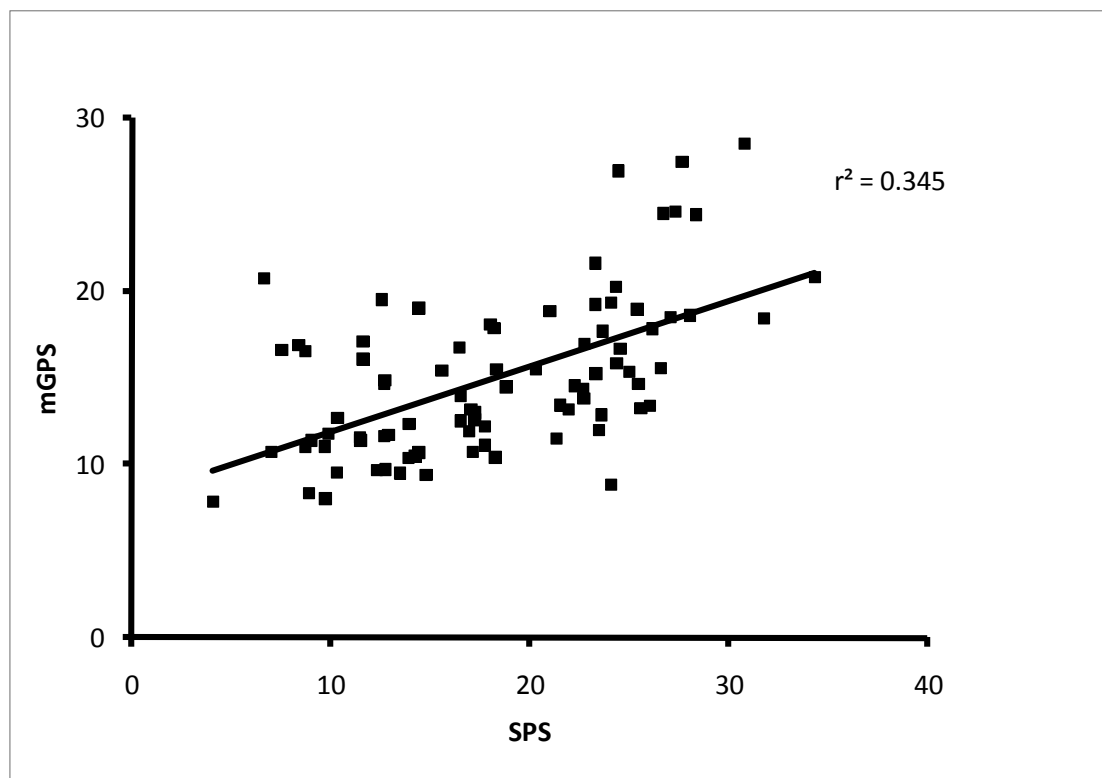
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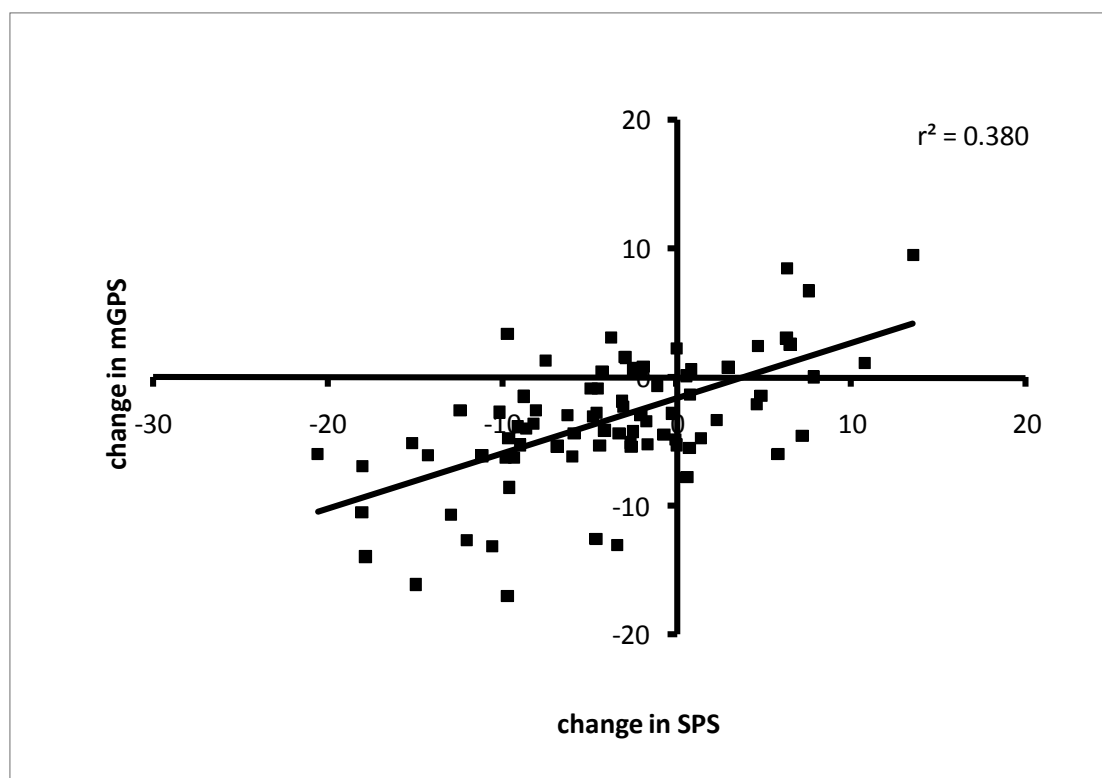


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Figure 1: Relationship between SPS and mGPS in pre-operative BSCP group

Figure 2: Relationship between change in SPS and mGPS in post-operative BSCP group





*Table 1: Timing of surgeries and assessments in young people with BSCP*

	Mean in Years	Range in years
Age at Surgery	10.5	4.8-19.4
Time from pre-op analysis to surgery	0.7	0.01-1.8
Time from surgery to post-op analysis	1.4	0.5-3.2

*Table 2: SPS and mGPS values pre- and post-operatively*

	Pre-Op	Post-Op	p-value
SPS	18.3° ±6.8°	15.0° ±4.6°	<0.001
mGPS	14.2° ±5.5°	11.7° ±3.8°	<0.001